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ANATOMIC VARIATIONS

A fusiform aneurysm of a persistent trigeminal artery variant: case report and literature review

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Abstract A 48-year-old man suffered from spontaneous subarachnoid hemorrhage. Emergent right internal carotid angiography showed the presence of a persistent trigeminal artery (PTA) variant with a fusiform aneurysm on its proximal segment where it branched from the internal carotid artery. This artery supplied the territory of the anterior inferior cerebellar artery. After consideration of the adequacy of the cerebellar circulation without this anomalous artery, intraluminal occlusion of the aneurysm together with the PTA variant was performed using detachable coils. The patient recovered uneventfully without any neurologic deficits.

Keywords Primitive trigeminal artery variant · Fusiform aneurysm · Endovascular treatment

Introduction

A persistent trigeminal artery (PTA) is a remnant of the embryonic circulation that connects the internal carotid and basilar arteries [8]. It has been reported to have an angiographic incidence of 0.1–0.2%. On very rare occasions, a variant form of the PTA connects the internal carotid to one of the cerebellar arteries instead of to the basilar artery. Aneurysms, though rare, can occur on either the PTA or its variant form, and are almost always saccular in form. Only two cases of fusiform aneurysm on this artery have been reported in the literature consulted [1, 6]. We here report a case of a fusiform aneurysm on a PTA variant, and its successful treatment with coils, and discuss how the anatomy determines the therapeutic options for the different types of PTA and aneurysm.

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Case report

A 48-year-old male with hypertension had been otherwise healthy until 11th November 2006, when he presented with sudden onset of severe headache, neck stiffness, nausea, vomiting, momentary loss of consciousness, and enuresis. On admission, a Hunt-Hess grade III subarachnoid hemorrhage was diagnosed.

Four-vessel cerebral digital subtraction angiography was performed and showed a $2.90 \times 3.53 \times 1.96$ mm^3 fusiform aneurysm in a variant artery that extended from the right internal carotid siphon and distributed to the territory normally supplied by the right anterior inferior cerebellar artery (AICA) (Fig. 1). Flow distal to the aneurysm was slow. The right posterior communicating artery was well developed, but the right vertebral artery was hypoplastic and supplied only the territory of the posterior inferior cerebellar artery (PICA). The left vertebral artery, in contrast, was well developed and supplied the basilar artery. The left AICA was also well developed and supplied the territory of the ipsilateral inferior cerebellum. The right AICA was hypoplastic and there was an opacification of a circumferential artery.

Before undertaking treatment, we considered the following observations. Distal flow from the PTA variant was minimal and very slow on angiography. The right AICA

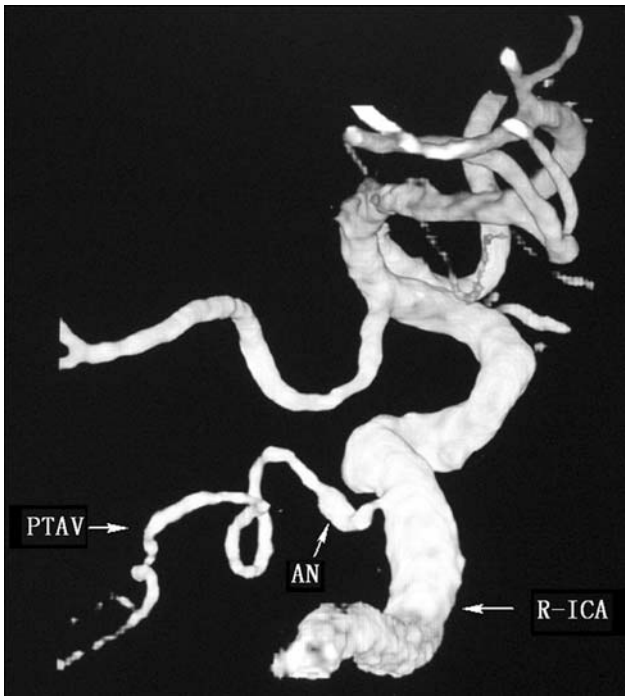

Fig. 1 The 3D reconstruction of right internal carotid artery (*R-ICA*) demonstrates the persistent trigeminal artery variant (*PTAV*) supplying the territory of cerebellum and the fusiform aneurysm (*AN*) on it

was present, but was hypoplastic. The patient, however, had no neurologic deficits caused by the ischemia of the AICA. The territory of the right PICA was supplied by the right vertebral artery, and the right superior cerebellar artery was normally developed. And most important to our consideration was the existence of flow, shown by an opacification, in a long circumferential artery. Considering the existence of a right AICA, the good flow in the other cerebellar arteries, and the existence of a potentially compensatory circumferential artery, we thought it safe to occlude the variant artery as well as its aneurysm.

Using a trans-femoral route, a Prowler-14 microcatheter (Cordis Therapeutics, Warren, NJ) was advanced coaxially through a 6F guiding catheter into the fusiform aneurysm. A Microplex-10 helical detachable coil (2 × 20 mm, MicroVention Inc, Aliso Viejo, CA) was advanced into the aneurysm. A control angiogram obtained just after the detachment showed slowed flow distal to the coil. A Hydrocoil 18 detachable coil (2 × 20 mm MicroVention Inc, Aliso Viejo, CA) and a second Microplex-10 helical coil (2 × 20 mm) were inserted into the aneurysm and the proximal end of the variant artery. The angiogram then showed only a small amount of contrast material in the aneurysm, and even less contrast material, as well as a slower flow, distal to the variant artery. Ten minutes later, an additional control angiography showed occlusion of the aneurysm and confirmed the obliteration of the variant artery. Also, the projection of the left vertebral artery showed that compensatory flow from the circumferential artery to the distal AICA was present.

The patient had no immediate neurologic deficits. His recovery was uneventful, and the occlusion of the variant artery as well as the establishment of collateral circulation was confirmed by the angiogram obtained before discharge.

Discussion

PTA variants have been classified in two ways. Uchino et al. [16] classified them as lateral and medial, depending on whether they originated in the posterolateral or posteromedial aspect of the cavernous internal carotid artery. Saltzman [10], on the other hand, classified them according to whether the anastomosis or the posterior communicating arteries supplied the posterior cerebral arteries. Saltzman types are the following: (1) the PTA supplies the posterior cerebral artery; (2) the PTA supplies one posterior cerebral artery and the posterior communicating artery supplies the other posterior cerebral artery; (3) the posterior communicating arteries supply both posterior cerebral arteries. The PTA variant described here would be classified as lateral (Uchino) and type 2 (Saltzman). The DSA image in the current case showed clearly that the AICA originated from the ICA. It might be considered as a very rare variation of the PTA similar to that previously described by Uchino et al. [14]. While the anomalous artery was occluded proximately, the distal AICA was supplied by the basilar artery via a "circumferential artery." The possibility that the "circumferential artery" was the small distal part of the PTA was excluded as DSA showed that the distal part of the PTA went to the cerebellum.

The persistent trigeminal artery is a remnant of the embryonic circulatory system. During embryonic development [8], it is present as an anastomosis between the aortic arch and one of the two longitudinal neural arteries. It begins to involute as the neural arteries fuse to become the basilar artery, and disappears by the time the embryo has reached a length of 14 mm.

Persistence of this embryonic artery is thought to occur as the result of incomplete fusion of some portion of the longitudinal neural arteries during the formation of the basilar artery, and results in a carotid–basilar anastomosis. But a rare variant can occur in which the trigeminal artery, instead, forms an anastomosis between the internal carotid artery and one of the cerebellar arteries.

Only 72 cases of such trigeminal artery variants have been described in the literature consulted (including our case) [3–5, 7, 11–13, 17]. Approximately 73% were reported to connect with the anterior inferior cerebellar artery, 13.5% with the superior cerebellar artery, and 13.5% with the posterior inferior cerebellar artery.

The prevalence of intracranial aneurysms in patients with PTA is about 3%, a prevalence similar to the prevalence of these aneurysms in the general population [2]. However, aneurysms on the PTA itself are rare [7] and only 44 cases have been reported in the literature consulted, 37 on the PTA and 7 [9, 10, 15] on the PTA variant. Most of these are saccular aneurysms, and only two fusiform aneurysms have been described [1, 6].

For a PTA aneurysm, it is important to determine to what extent the PTA supplies the blood flow to the distal portion of the basilar, superior cerebellum, and posterior cerebral artery territories. Autopsy reports of adult cases of PTA have even described two arterial branches originating from its cisternal portion: an artery supplying the trigeminal nerve root and a branch sending a perforating artery directly into the pons [9, 12]. Pontine branches are most probably the functioning vessels supplying the brain stem, and therefore occlusion of the PTA, if such branches are present, may cause ischemic lesions in the brain stem.

For a saccular aneurysm, either open surgery or interventional techniques can produce a satisfactory result, but in the case of fusiform aneurysm, direct surgical clipping of the aneurysm while at the same time preserving the patency of PTA is extremely difficult.

For a fusiform aneurysm on a PTA variant, to occlude the aneurysm and to also preserve the artery is impossible. To avoid re-bleeding, the PTA variant needs to be occluded. Unlike cases involving a PTA, when a PTA variant is present, the basilar artery is always normally developed. Therefore, it is always able to provide the brain stem with a good blood supply. As for the cerebellar blood supply, if the arteries supplying the adjacent territory have a normal blood supply, occluding the PTA variant will be safe. In rare cases, if prior deficits exist in the posterior circulation, no sufficient collateral circulation will be available to compensate for the loss of the PTA variant. In this circumstance, a bypass operation should be performed before the parent artery's occlusion.

Although PTA variants are rare, because most cases of PTA variants reported in the literature were diagnosed due to the hemorrhage of the anomalous artery, the prevalence of this anatomic variation may be underestimated. It has been demonstrated that MR angiography can detect this variation [15], and, as a non-invasive method, MR angiography could be applied in the future in a screening study of PTA variants. Such a screening study may reveal the true prevalence of this anomaly in the population.

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